

Complex regional pain syndrome (CRPS) type-1 following snake bite: a case report

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ABSTRACT

The pathophysiological mechanism and clinical course of complex regional pain syndrome (CRPS) type-1 still remain ill defined. Both the treatment and the prediction of the outcome of the treatment are difficult. Abnormal neurohumoral and inflammatory mechanisms have been implicated in its causation usually following trivial noxious event in an extremity. However, to the best of our knowledge CRPS type-1 following snakebite has not been reported yet in the literature. We here report a case of an aggressive CRPS type-1 following a mountain pit viper bite, locally known as *Gurube* (*Ovophis monticola monticola*) in a 55-year-old lady. The clinical condition responded well to the therapy with serial sympathetic blockade of the limb with local anaesthetics, non-steroidal antiinflammatory analgesic, antiepileptic, antidepressant and physiotherapy. Our experience in managing this patient and associated pathophysiology in development of CRPS type-1 are discussed.

Keywords: snake bite, local toxicity, complex regional pain syndrome type-1, pain, sympathetic block

Complex regional pain syndrome (CRPS) type-1 is a chronic pain syndrome involving limbs. It usually develops following a trivial noxious event. It is characterized by edema, changes in the blood flow, abnormal sudomotor activity (sweating) in the region of the pain, or allodynia.¹ The triggered vicious cycle of pain and sympathetic overactivity leads to osteoporosis in the affected limb.² Increased blood flow to the bone consequent to alteration of haemodynamics has been attributed to the development of osteoporosis.³ Etiological factors described include trauma, infection, neoplasm, systemic diseases and many other non-specific conditions including inflammation and immobility.⁴ However, CRPS type-1 following snakebite has not yet been reported in the literature. Management of a patient who developed CRPS type-1 following snakebite and the associated pathophysiology in the development of same has been presented here.

CASE REPORT

A 55-year-old lady from Phurumba Village Development Committee of remote mountainous Taplejung District of Nepal presented to the orthopedic department of our hospital with complaints of pain and swelling of her left hand. She was then referred to our pain clinic for further management. She gives history of bite by a snake locally known as *Gurube Sarpa* (*Ovophis monticola monticola*) on her left ring finger 3 weeks prior to presentation to the pain clinic. Following the bite patient had severe burning pain and swelling of the left hand. A tourniquet was applied initially but was removed within one hour.

She took local herbal treatment (nature not known) topically along with oral ibuprofen for pain relief but did not receive any anti snake venom due to unavailability. The affected hand became darkish brown in color without any perceptible pain relief. Although the swelling decreased to start but its colour worsened from darkish brown to greyish. Pain in hand, however, persisted to restrict the mobility of her hand. In 2 weeks time after the bite, the stiffness of the wrist and other small joints of the hand worsened and the hand became tender to even slightest touch. The pain started to ascend to the proximal part of the limb. The hand remained swollen and the skin became shiny and cold. She could not move her limb due to pain. She felt anxious and her sleep was disturbed by the pain.

On examination the skin of the affected hand was pale, greyish in colour with shiny texture. It was oedematous (non-pitting), cold, moist and tender to touch. The wrist and other small joints of the limb were stiff.

X-ray of the hand showed demineralization of the bones of the hand (Fig.1). Biochemical and haematological test results were within normal limits.

Based on these findings, the provisional diagnosis of CRPS type-1 was made and the treatment modality was planned. Patient's left stellate ganglion was blocked with injection of 8 ml of 0.5% bupivacaine using anterior approach. The patient was given oral diclofenac 50mg twice daily, amitriptyline 25mg once daily and capsule gabapentin 300mg twice daily along with limb



Fig. 1. Plain X-ray of the hands showing significant demineralization in the small bones of left hand. Significant soft tissue swelling is also evident



Fig. 2. Plain X-ray of the hands showing improved bone density of the small bones of the left hand after the treatment. Soft tissue swelling has also reduced

physiotherapy. Stellate ganglion blocks were given 3 times a week until 10 blocks were completed. We continued oral medications and physiotherapy in this period. The patient responded to this treatment with a significant decrease in pain intensity from 8 to 2 in 0-10 numerical analogue scale (zero indicating no pain, 10 indicating maximum imaginable pain) and complete functional recovery. Repeat X-ray showed significant improvement in the bone density (Fig.2) and reduction in the soft tissue swelling. The patient was advised to continue oral medication and physiotherapy and come for follow up after one month. The patient is in follow up.

DISCUSSION

CRPS type-1 is one of the most difficult and challenging condition encountered by a pain physician. It is often initiated by trivial trauma that the patient may not even remember.⁴ Although how CRPS type-1 progresses to such a destructive state (often leading to trophic changes) is not clear, it is attributed to the alteration in the peripheral autonomic nervous system response and subsequently to a vicious cycle of pain and sympathetic over activity.² Sympathetic dysfunction along with disuse of the limb may result in bone demineralization.⁴

In our case, the inciting event was bite by a locally poisonous snake locally known as *Gurube* (*Ovophis monticola monticola* or Mountain pit viper). We could not find any case report of CRPS type-1 incited by snake bite in PubMed® (National Library of Medicine, USA) search. It is well described that CRPS type-1 may be initiated by any trivial injury in the periphery, but rapid course in our case evidenced both by clinical and radiological findings compell us to speculate that snake venom can play a role in causation or exaggeration of CRPS type-1. Moreover, to add to the complexity, local

swelling and pain due to snake bite may mask the progressing CRPS type-1. Local toxicities of viperid and crotalid species are characterized by local inflammation, pain and tissue damage.⁵ Phospholipase A2 and metalloproteinase along with inflammatory substances such as interleukins and tumor necrosis factors have been implicated for the type of local tissue effects.⁶ Although no anti snake venom was used in our case, anti snake venoms even if used are not satisfactorily effective in preventing local tissue damage and inflammation.⁷ Further, presence of various inflammatory substances (such as Interleukin-6 and Tumour Necrosis Factor-alpha) in the affected limb in CRPS type-1 patients have demonstrated inflammatory nature of the disease⁸⁻¹⁰ and the inflammatory substances present in the snake venom must have played a role in the course of the disease in our case. It is understandable that multitude of variables in both the etiologic event and the subsequent disease makes it very difficult to establish straight forward cause effect relationship in a complex case of CRPS type-1. Despite these facts, typical clinical and radiological findings and rapid response to sympathetic blockade in our case confirms the diagnosis of CRPS type-1 and demonstrates a strong possibility of association between snakebite and aggressive form of CRPS type-1. We believe that this case will be of value for the interested researchers in formulating hypothesis in further understanding of the pathophysiology of CRPS type-1.

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